

Review

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## Appendiceal Intussusception

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**ABSTRACT** The pathology of the appendix remains one of the topical areas of abdominal surgery. Its rare diseases can be difficult to diagnose and treat, and are accompanied by the development of serious complications. One of them is intussusception of the appendix to the cecum. Intussusception can be caused by previously undiagnosed primary lesions of the appendix. At the same time, it can simulate tumor and tumor-like formations of the cecum and lead to diagnostic errors. The literature review provides modern data on the history of the study of the problem, the epidemiology of classification, the causes and mechanisms of the development of appendiceal intussusception, the methods of its diagnosis and treatment.

**Keywords:** appendix, intussusception, appendicitis, cecum, tumor, appendectomy

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VAI – vermiform appendix invagination

VA – vermiform appendix

### INTRODUCTION.

Vermiform appendix invagination (VAI) as the primary problem connected with anatomic features of the vermiform appendix (VA), its motor activity, as well as the secondary problem connected with the number of benign neoplasms and malignant formations in the VA itself and ileocecal area, is not enough studied and highlighted in the medical literature.

According to the P.F. Kaliteevskiy expression “vermiform appendix non-inflammatory diseases have vanished in the flow of appendicitis» (1). However along with different forms of appendicitis, benign neoplasms and malignant formations, parasitic diseases there is a number of pathologic processes that may lead to serious complications and are of great difficulty for pre-operational diagnostics and surgical treatment tactics choice.

Authors did not aim at choosing all publications from all available sources depicting single VAI, the review is based on previously published summarized data depicting not less than 5 cases.

## EPIDEMIOLOGY, ETIOLOGY AND PATHOGENESIS

Intussusception of the appendix is a rather rare surgical pathology. According to the available domestic and foreign literature, among all cases of intestinal obstruction, VAI occurs in 3-4% [1-11].

The invagination of the appendix was first described by McKidd in 1858 as an autopsy finding in a seven-year-old boy [12]. In the next century and a half, publications with a description of single or small series (no more than 5 cases) predominate, and most of the publications highlight cases of associated (secondary) VAI associated with the presence of various pathologies, including tumors of the VA [13-21].

Publications on intussusception of a "healthy" appendix by the type "into itself" are sporadic [2-5, 7-10, 12, 22-25].

In this regard, a clear separation of primary and secondary intussusceptions is necessary.

The etiology of primary VAIs in most cases remains unknown. However, some features have been identified that predispose to the development of intussusception; they are represented by two groups: anatomical and pathological [1, 3].

Anatomical conditions:

- fetal type of cecum with an appendix in the region of the apex of its dome;
- an increase in the diameter of the lumen of the appendix with a large lumen of the proximal part in comparison with the distal one;
- a thin mesentery of the VA with a reduced volume of fiber and a narrow base.

Pathological conditions are associated with abnormal peristalsis of the appendix due to its irritation. The ability to self-peristalsis of the VA was radiologically determined by S.P. Grigoriev [1]. Its longitudinal musculature reacts to acetylcholine with tonic contractions, annular - periodic, while the longitudinal is more powerful and excitable. Thus, both intramural and intraluminal stimulants irritate and activate the VA peristalsis, which leads to the appearance of the intussusception onset point in a certain part of the wall, pulling the rest of the VA. Such areas are called "leading points", which are then retracted into the lumen of the VA or the dome of the cecum by peristalsis, which is the main mechanism of primary intussusception [1, 11].

Secondary intussusceptions are associated with the presence of a pathological focus in the VA itself, which is the leading factor [3, 11, 15, 16, 19, 20, 26-29].

Tumors of the VA amount to 0.5% of all tumors of the gastrointestinal tract, half of them are described in the literature as the leading item in the AVI [3, 11, 15, 19, 20].

For the first time, adenocarcinoma of VA was described by Berger (1882), endometriosis - by K. Rokitansky (1860), and in Russia - by F.K. Weber (1907) [1].

In 1963, D. Collins published the results of a study of 71 thousand observations of various pathologies of VA for 40 years, while the frequency of intussusceptions was 0.01%, endometriosis as the leading item was noted in 0.05%, adenocarcinoma of VA in 0.08% of cases [6].

Costa et al. in 2014 analyzed 194 secondary VAI cases published in the PubMed database from 1925 to 2012 using the keywords "appendix intussusception", "appendiceal intussusception", "intussuscepted AND appendix" and "appendiceal AND intussuscepted" [30].

In the most complete, of the latest literary reviews (2017), devoted to a greater extent to secondary VAIs, it is noted that most often (28%) the role of the leading point of intussusception is played by acute inflammation of the VA (acute appendicitis) [11].

The next common cause was endometriosis, which was diagnosed in 23.6% of patients. Mucocele as the leading point of VAI was observed in 15.9%, mucinous cystadenomas - in 4.4%, adenomas - in 7.7%, adenocarcinomas - in 4.4%, carcinoid tumors - in 4.9% and in one case (0.55%) diagnosed with neuroendocrine cancer with metastases to the lymph nodes.

Other tumors of the invaginated appendix are, in one example, papilloma, hamartoma, juvenile polyp, MALT lymphoma, and ileocecal non-Hodgkin's lymphoma. In total, 74 patients (40.6%) had benign or malignant tumors. In this study, on the basis of 220 cases of VAI described in the literature, its gender characteristics are given, men accounted for 37.6%, women - 62.4%. Pediatric cases accounted for 23.8%, adult patients - 76.2% [11].

The classification and types of VAI proposed by V. McSwain in 1941, with some additions, are still relevant and generally accepted [12].

Type I: the apex of the appendix forms the invaginate and penetrates into its proximal part.

Type II: intussusception begins in some part of the appendix. The cavity for the invaginate is formed by the adjacent tissues.

Type III: intussusception begins at the junction of the appendix with the cecum [31].

Type IV: retrograde invagination, in which the proximal part is invaginated into the distal one.

Type V: complete intussusception as a result of progression of type 1, type 2, or type 3 intussusception.

"itself to itself" intussusception is less common than others [1–4]. D. Collins, as a result of a study of 71 thousand VA after appendectomy, only in one case revealed this type of VAI [6]. P.F. Kaliteevsky and K.S. Rukosuev in the 70s. of the last century, when describing their VAI observations report five published cases of "itself to itself" VAI [1, 4]. In the available literature for 10 years, we found 14 descriptions of this type of invagination [2–5, 7, 9, 10, 12, 15, 22–25, 32].

Langsam et al. in 1984 described in detail the mechanisms of formation of an isolated VAI, taking into account the localization of the leading point [33]:

1. The head of the invaginate is the apex of the appendix:

1.1. The head of the invaginate is introduced into the lumen of the appendix to different depths, not reaching its base.

1.2. The head of the invaginate is inserted up to the cavity of the cecum. In this case, as in the previous one, the apex of the appendix is in the invaginate, the size of the visible part of the VA decreases.

1.3. The head of the invaginate, moving forward, is introduced into the cavity of the cecum, while the appendix is turned out like a finger of a glove.

The dome of the cecum, as a rule, is not involved in invaginate.

The formation of a small funnel-shaped retraction of the dome of the cecum in the circumference of the base of the appendix is possible.

2. The head of the invaginate is the proximal part of the appendix:

2.1. The head of the invaginate is located closer to the base of the VA, while the apex of the appendix remains outside the invaginate.

2.2. With deep penetration of the proximal part of the appendix, the VA can completely disappear into the invaginate.

2.3. When the VA is introduced from the base, the head of the invaginate immediately appears in the cavity of the cecum, and the apex of the appendix may remain outside the invaginate.

2.4. With further advancement of the head of the invaginate, the VA may completely appear in the lumen of the invaginate. In this case, the dome of the cecum in the circumference of the base of the appendix is often drawn into the cavity of the cecum, forming a funnel-shaped retraction.

With all variants of the introduction of VA through its lumen into the cavity of the cecum, it is turned by the mucous membrane outward like a glove finger and is located in the cecum like a bell tongue. In other types of intussusception (ileocecal, blind-colon), the process is passively involved in the invaginate and remains non-inverted [33].

## DIAGNOSTICS

Preoperative diagnosis of VAI is extremely difficult, and the diagnosis, as a rule, is intraoperative when VAI is combined with destructive appendicitis [1, 3–5, 7, 10, 20, 22, 32, 34, 35].

It is also difficult to determine the cause and type of VAI preoperatively, although the tactics and volume of surgical intervention depend on this.

However, in the publication of Tsukamoto R. et al., in 32% of cases before the operation, a disease was detected, which led to a secondary VAI [36].

Ultrasonography plays an important role as a diagnostic tool for VAI. Sonographic features include multiple concentric hyperechoic and hypoechoic rings. Ultrasound examination of the abdominal cavity allows to identify the "target feature", the concentric ring as part of the appendix topography, which practically confirms the diagnosis of VAI [35, 37].

Visualization of the VA pathology during irrigoscopy plays an important role, however, anatomical features and nonspecific radiological symptoms can make it difficult to accurately determine the presence or absence of VA pathology. A number of studies have noted that the appearance of a "twisted spring" in the dome of the cecum with non-filling of the lumen of the VA with tight filling of the cecum with barium suspension is a sign of a clear or proven VAI [30, 37, 38].

Computed tomography of the abdominal cavity is the most commonly used method for diagnosing VAI. The wall of the cecum, due to repeated invaginations, undergoes inflammation and fibrosis, which leads to the formation of a mass-like formation. The presence of a concentric central mass, a "target sign," having a layered, sausage-like or kidney-like appearance, is an almost pathognomonic sign of VAI in computer diagnostics [30, 35, 37, 38].

Colonoscopy allows direct visualization of the lesion, while the invaginated appendix looks like a polypoid formation covered with a normal mucous membrane, in the form of an adenomatous polyp with a central retraction in the anatomical place of the VA, or a long invaginated tubular structure ("bell tongue") protruding from the appendicular opening into the lumen of the cecum [39, 40].

## TREATMENT

The invagination of the appendix in 63% has a chronic form, casuistic cases of spontaneous disinvagination of the VA have been described, there are reports of regression of invagination and clinical symptoms after a barium enema. Nevertheless, the risk of recurrence is high and most surgeons advocate surgical treatment [1, 9, 41].

Some surgeons recommend appendectomy in conjunction with resection of the caecum cuff around the base of the VA. In their opinion, the advantage of this method is to eliminate the risk of failure of the VA stump, resection of the cecum cuff provides reliable closure of the resulting wound defect of its dome.

Since most lesions of the VA are benign, cuff resection together with appendectomy provides complete cure [1, 9, 21, 35, 37, 38, 41–43].

Not a single method for diagnosing ICO currently allows solving the main question, whether the process is benign or malignant, since intussusception forms a tumor mass, its malignancy cannot be determined even during surgery. Until now, the basic principle of choosing the scope of the operation is the experience and oncological vigilance of the operating surgeon. If intussusception is misdiagnosed as an VA neoplasm before or during surgery, surgical treatment is performed as if the process is a malignant VA neoplasm.

If a mucous cyst or adenoma is the cause, appendectomy is appropriate. Carcinoid tumors and adenocarcinoma require right-sided hemicolectomy with lymphadenectomy [35, 36, 40].

There are no clear recommendations in the literature on the choice of the scope of surgical treatment.

Appendectomy is acceptable if the tumor process is located within the VA and can be completely removed. Right-sided hemicolectomy should be performed when the tumor infiltrate has spread to the dome of the cecum [37, 39, 41, 44].

According to Chaar C.I. et al. (2009), 49% of patients underwent simple appendectomy, 48% of patients underwent extended operations with resection of the dome of the cecum or hemicolectomy, laparoscopic appendectomy was performed in 2% of cases [5].

Several case studies have reported the colonoscopic appendectomy using the Endoloop ligature [39, 40].

In a later study by Soylu L. et al. (2014) for the treatment of VAI : ileocecectomy (27%), right-sided hemicolectomy (21%) and subtotal colectomy (1%) are used. Colonoscopic appendectomy was performed in 4 patients (3%). This method is effective and safe if the endoscopist detects intussusception of the appendix without signs of tumor lesion [28].

The use of laparoscopic VAI surgery for tumor lesions of the VA remains a controversial issue. The main problem to be solved in this case is the adequacy of the resection volume depending on the spread of the tumor, especially in urgent surgery [39, 41].

In this regard, the reports that have appeared since the beginning of 2000 about significant advances in endoscopic, radiation and laparoscopic diagnostics of asymptomatic VAI deserve attention. Patients underwent various kinds of examinations in connection with recurrent non-localized pain in the abdomen, with transient disturbances of intestinal passage, or the presence of VAI was a diagnostic finding [2, 5, 8, 13-15, 17-19, 21, 24, 25, 32, 38, 43].

Expansion of diagnostic capabilities, the absence of the need for an emergency or urgent operation can make it possible to verify the nature of the process and select an adequate amount of operational aid.

## CONCLUSION

Thus, the epidemiology of the vermiform appendix invagination based on intra-operative and in rare cases pre-operative diagnostics is doubtful, the spread of vermiform appendix invagination is much higher as in many cases it is asymptomatic.

Vermiform appendix invagination present variants and possibility of their combinations particularly during the asymptomatic course are difficult to diagnose and surgically treat in due time.

The material analysis makes us conclude that the modern diagnostics issue and particularly the problem of the adequate surgical vermiform appendix invagination treatment is surgery-relevant and requires tactics change. Most often the vermiform appendix invagination is the secondary pathology, associated with the primary ones including malignant diseases of the vermiform appendix. Both intra-operative diagnostics accuracy, and the degree of the tumor process spread impact the surgery tactics.

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